

The development and validation of the Epilepsy and Learning Disability Quality of Life (ELDQOL) scale

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Abstract

Few suitable instruments exist for use with people, especially children, with both epilepsy and learning disabilities. One such measure is the Epilepsy and Learning Disabilities Quality of Life scale (ELDQOL), which has recently undergone revision following feedback from relevant users. This paper reports on the final psychometric testing phase of ELDQOL. ELDQOL consists of 70 items covering seizure severity, seizure-related injuries, AED side-effects, behaviour, mood, physical, cognitive and social functioning, parental concern, communication, overall QOL and overall health. Revalidation involved a qualitative phase to ascertain users' opinions on the wording, coverage and layout of the questionnaire; and a quantitative phase to examine internal consistency and test-retest reliability, and validity. The final version of ELDQOL has very good evidence of reliability and validity, making it a promising instrument for assessing QOL in children/young adults with epilepsy and learning disability.

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Introduction

There is general agreement within the health outcomes field of the importance of assessing therapeutic outcomes including quality of life (QOL).¹ Medical interventions may be beneficial to patients on impairment or disability measures, but without equally refined QOL measurements a clear and comprehensive evaluation of their efficacy is not possible.^{2,3} The assessment of QOL in adults with epilepsy is now widespread with the existence of a number of psychometrically sound measures.⁴ However, reviews of outcome measures have shown that few suitable instruments currently exist for use with people who have both epilepsy and learning disabilities.^{1,5}

One recently developed measure, the Epilepsy Outcome Scale (EOS),⁶ was developed to be completed by carers of people with epilepsy and learning disabilities (LD), but while valid and reliable, it is intended for adults rather than children with these conditions. A recent review of QOL measures for use specifically with children and adolescents with epilepsy¹ identified five epilepsy-specific instruments, of which three were considered potentially suitable for children who had LD: the Health-related Quality of Life in Children with Epilepsy measure;⁷ the Impact of Childhood Neurological Disability Scale (ICND);⁸ and the Epilepsy and Learning Disabilities Quality of Life scale (ELDQOL).⁹⁻¹²

The Health-related Quality of Life in Children with Epilepsy measure was not purposely developed for those with both epilepsy and LD. However, it does incorporate a self-report (completed by the child) as well as a proxy-report which can be completed by parents alongside or instead of the child report, and seems to be reasonably valid and reliable.¹ The ICND assesses the impact of epilepsy and concomitant behavioural, cognitive, and physical/neurological disability in children. It has good reliability and is reported to be satisfactory in terms of validity.¹

ELDQOL is specifically aimed at informal/formal carers of children with both severe epilepsy and LD. Initial evidence of its psychometric properties was reported by the scale's developers¹³, and subsequently ELDQOL was reviewed by Espie et al⁵ who also reported evidence of internal and test-retest reliability, and some evidence of sensitivity. ELDQOL has been used in two studies, results of which have been

reported.^{11,14}

However, as a result of feedback from clinicians involved, it was decided to undertake revision and revalidation of ELDQOL. This paper reports on the psychometric testing of the final, revised version of ELDQOL. A detailed report of the earlier stages in its development is available from one of the authors (AJ). A summary of its history will be provided here.

Initial development of ELDQOL

Potential items for inclusion in the pilot structured measure were selected based on in-depth interviews with parents of children with severe epilepsy. The measure was then piloted to assess its psychometric properties (content and construct validity, internal consistency and test-retest reliability) and acceptability to parents. Parents of 50 patients across 5 UK paediatric neurology out-patient departments were recruited to the pilot study.

The pilot version of ELDQOL consisted of 66 items across 6 domains: seizure severity, seizure-related injuries, AED side-effects, behaviour, mood and overall QOL. It was found to be satisfactory in terms of content and construct validity and internal consistency, and showed high test-retest reliability. Furthermore, parents commented that the measure was comprehensive, relevant and easy to complete. The pilot version of ELDQOL was subsequently used in a clinical trial of Lamictal¹¹ which provided evidence of its responsiveness. However, a number of criticisms were made about the measure by clinicians involved in the trials, particularly in terms of the clarity of meaning of several items and of the appropriateness or sensitivity of response sets.

Consequently, a revalidation study was undertaken to pre-test (using cognitive interviews¹⁵), revise and then reassess the psychometric properties of ELDQOL. The purpose of this paper is to describe this final phase in the development of the measure and report its psychometric properties.

Methods

Qualitative phase

Qualitative (cognitive) interviews were undertaken with 16 parents and 17 health and other professionals in order to ascertain their opinions on the wording, coverage and layout of the questionnaire.

The parent participant group comprised 5 couples (the young person's mother and father) and 6 mothers. The 11 children of these parents were 8 boys and 3 girls, age range 18 months to 21 years. Two young people attended residential school, 7 attended special schools or day centres, and one a mainstream school. The parents were identified by a consultant or senior nurse at 2 specialist hospitals in the north of England. They were selected because of their child's condition and because they were judged to be interested in all aspects of their care.

The professional participant group consisted of 10 consultant paediatric neurologists or neurologists with a special interest in epilepsy and learning disabilities, one associated health services researcher, two paediatric epilepsy nurse specialists, two nurses specialising in epilepsy and learning disabilities, two teachers/play leaders, and one psychologist.

All participants were sent a copy of the existing ELDQOL questionnaire and asked to complete it immediately prior to the in-depth interview. Interviews were tape recorded and lasted approximately one hour. They involved scrutiny of each question in turn, where participants' views were sought on interpretation, coverage, content, wording and scale construction. The tapes were transcribed for analysis.

Psychometric phase

Following revision based on the findings of the cognitive interviews, the final version of ELDQOL was administered in a postal survey to 47 parents/guardians and 21 formal carers of children with epilepsy and learning disabilities. The parents were identified by consultant paediatric neurologists and epilepsy specialist nurses at 4 UK hospitals,

who explained the study and asked whether they would be willing to complete the questionnaire. Those who agreed were sent a study information sheet together with a copy of the questionnaire and a consent form and asked to return both when they had been completed. For the purposes of evaluating test-retest reliability, parents were sent a second questionnaire approximately 2 weeks after completing the first. Formal carers were identified through a national long-stay residential/assessment centre.

The mean age of children recruited to the study was 11.5 years (SD 4.6, range 2 – 19¹); 58% were male and 42% female. Of the parent respondents, 39 were mothers, 3 were fathers, and 2 were other relatives; 3 of the questionnaires were jointly completed by both parents.

The final version consisted of 70 items covering seizure severity, seizure-related injuries, AED side-effects, behaviour, mood, physical functioning, cognitive functioning, social functioning, parental concern, communication, overall QOL and overall health. The items covering behaviour, seizure severity, mood and side-effects domains are summed to create 4 subscales containing 9, 14, 16 and 19 items respectively. On each subscale, a higher score indicates poorer functioning. The remaining domains consist of single or several non-summed items. Subscales from two other outcome measures [the Aberrant Behavior Checklist (ABC)¹⁶ and the Child Health Questionnaire (CHQ)]¹⁷ were also administered for the purposes of evaluating the construct validity of ELDQOL.

In order to evaluate validity, t-tests and correlations were used to examine the relationship between ELDQOL sub-scales and:

- * the Irritability and Hyperactivity subscales of the ABC;
- * the Impact on Parental Time, Emotional Impact on Parent, and Family Activities sub-scales of the CHQ;
- * 4 global items (overall health, overall QOL, perceived severity of condition and

¹ Age was not recorded for 13 children.

disability level).

Severity of disability was assessed using a 4-item scale developed by a consultant paediatric neurologist (RA) with a specialist interest in epilepsy and learning disability. It covers mobility, feeding, dressing and speech, each item having 5 response options ranging from the worst to the best possible level of functioning.

It was predicted a priori that there would be:

- * a significant relationship between the ABC Irritability and Hyperactivity scales and scores on each ELDQOL subscale;
- * a significant relationship between the 3 CHQ scales and scores on each ELDQOL subscale;
- * a significant relationship between overall health and scores on each ELDQOL subscale;
- * A significant relationship between overall QOL and scores on the Side-Effects, Seizure Severity, and Mood subscales
- * A significant relationship between perceived severity of condition and each subscale
- * A significant relationship between disability level and Seizure Severity.

Internal consistency of the 4 sub-scales was assessed using Cronbach's alpha coefficients and item-total correlations. Test-retest reliability was assessed using intraclass correlation coefficients. Floor and ceiling effects for each subscale were examined.

Results

Qualitative phase

Views of parents

Six issues were identified through the cognitive interviews with parents: validity of domains, multiple seizure types, wording, ease of completion, response categories and identifying change. It was clear that the questionnaire resonated with parents' experiences and concerns, the questions being described as 'very good' and 'geared to parents' for example. However, it did not, in its existing form, take into account that some children have no mobility or verbal skills. Some commented on the lack of lifestyle questions to capture social activities and interests. In terms of multiple seizure types, some parents found it difficult to identify which seizure type they should refer to in answering the questions. There was a consensus that use of the word 'seizure' was preferable to 'attack'. The 'layman's language' was appreciated because it was easy to understand, and the length of the questionnaire was viewed as good, 'quick to do' and 'easily to fill in'. Instructions were described as 'clear', 'logical' and 'concise'. Overall, parents felt that they had sufficient choice in response categories, and being asked to concentrate on the past 4 weeks seemed acceptable.

Views of professionals

Interviews with professionals highlighted several broad methodological issues, in particular the nature of the proxy respondent, scope and objectives of the questionnaire, sensitivity to change, and questionnaire design. Participants felt generally that parents would be able to answer the majority of the questions and would therefore be appropriate proxies. However, there was some reservation regarding the suitability of nursing, teaching or respite care staff who may not have known the child for long – '*it would be necessary to know the child inside out*' to be able to complete some of the sections of the questionnaire.. In terms of the scope and objectives of the questionnaire, its orientation led some to query what was being sought and by whom it would be used. The addition of socially based QOL questions, a 'health profile' and a section covering function was suggested. In terms of design, most professionals felt the length of the

questionnaire was about right. Some thought the use of the word ‘attack’ was regressive in terms of practice and preferred the term ‘seizure’.

Revisions made as a result of the qualitative phase

Taking into account the comments from parents and professionals, a number of revisions were made. In summary:

- * the word ‘seizures’ replaced the word ‘attacks’
- * ‘can’t say’ and ‘does not apply’ response options were added
- * a question about perceived control was dropped because of ambiguity of meaning; and one about perceived severity was added
- * an item was added to those about seizure-related injuries
- * an item covering ‘usual activities’ (school, playing, socialising) was added to the behaviour subscale
- * 2 items (‘sociable’ and ‘depressed’) were dropped from the mood subscale and replaced by three new ones (‘sad’, ‘withdrawn’, ‘cooperative/helpful’)
- * 2 items were dropped from the side effects profile (hair loss, dizziness) and 2 were added (weight loss, loss of appetite); and a single item, ‘restlessness/hyperactivity’ was separated into two
- * clearer/more detailed instructions were provided (including a statement instructing parents/proxies to think about the most severe seizures in the case of multiple seizure types, and emphasising that questions relate to the last four weeks)

Psychometric phase

Reliability

As shown in Table 1, internal consistency of the Behaviour, Seizure Severity, Mood and Side-Effects scales was high (0.74 – 0.95). Item-total correlations exceeded the desired 0.4 for most items, although in each scale there were some items which did not meet this: 3/9 items in the Behaviour Scale ('problems sleeping', 'being prevented from taking part in normal activities', and 'appetite a problem'); 7/14 items in the Seizure Severity scale ('aware of surroundings after seizure', 'blank out/lose consciousness', 'fall to the ground', 'soil self', 'injure mouth/cheek /tongue', 'upset by injuries', 'appear sleepy/subdued'); 4/16 items in the Mood scale ('tearful', 'hyperactive', 'withdrawn', 'cooperative'); and 3/19 items in the Side-effects scale ('skin problems', 'shaky hands', 'weight gain).

The test-retest reliability of each subscale was high (range 0.80 - 0.96, Table 1). Floor/ceiling effects were acceptable in the Side Effects scale, low in the Seizure Severity scale, and there were none in the Mood or Behaviour scales (Table 1).

Validity

Table 2 shows that all but one of the 4 ELDQOL subscales (Behaviour) were moderately to highly correlated with both the Irritability and Hyperactivity scales of the ABC, and that all 4 were moderately to highly correlated with the Emotional Impact and Family Activities scales of the CHQ. Two of the ELDQOL subscales (Seizure Severity and Side-effects) correlated moderately with the Impact on Parental Time scale of the CHQ.

Table 3 shows a significant relationship between mean scores on the Behaviour and Mood subscales by overall health. There was a similar though non-significant trend for the Seizure Severity and Side Effects subscales.

As can be seen in Table 4, those with poorer perceived overall QOL tended to have higher mean scores on the ELDQOL scales, although these were not statistically significant.

Mean scores on each of the 4 subscales were significantly worse for parents who felt their child's condition was very severe compared to those who felt it was somewhat or moderately severe (Table 5).

The Seizure Severity and Behaviour subscales were significantly correlated with severity of disability (Table 6). No significant correlation was found for the Side-Effects and Mood subscales.

Discussion

ELDQOL was derived using currently accepted standards for scale development, including qualitative and cognitive interview techniques with the target groups. The ongoing involvement of parents and professionals in determining the content and format of the measure has enhanced its acceptability and ease of completion. The final version of ELDQOL has very good evidence of reliability and validity, making it a promising proxy instrument for assessing the QOL of children/young adults with epilepsy and learning disability. Although in this validation exercise parents were not asked to note length of time taken to complete ELDQOL, this was previously estimated as only around 20 minutes⁵, a factor which may further contribute to its acceptability.

In terms of internal consistency, each of the 4 subscales had high Cronbach's alpha values. For each scale there were some items which did not meet the desired 0.4 level for item to total scale correlation.¹⁹ However, in each case the effect on Cronbach's alpha when these items are removed from the scale is negligible or has no effect and we therefore propose to retain these items on the grounds of comprehensiveness/content validity.

The test-retest exercise found that each of the 4 subscales achieved ICCs well above the required level of 0.70 for group comparisons, and 3 of the 4 performed well enough for comparing individuals over time (0.90).¹⁹ Each scale performed well or perfectly in terms of floor/ceiling effects.

We found excellent evidence of construct validity as all but one of the 4 ELDQOL subscales (Behaviour) were moderately to highly correlated with both the Irritability and Hyperactivity scales of the ABC, and all were moderately to highly correlated with the Emotional Impact and Family Activities scales of the CHQ. Poorer perceived health was significantly related to poorer mean scores on two of the ELDQOL subscales, and a similar trend was seen for the remaining two, providing further evidence of construct validity. Greater perceived severity of the condition was significantly related to higher scores on each subscale, and disability level was significantly correlated with scores on the Seizure Severity and Behaviour scales.

Although the sample size was relatively small we would suggest that, based on both psychometric analysis and feedback from parents, ELDQOL will be a valuable contribution to outcomes measurement in childhood epilepsy and learning disability, particularly in the context of treatment trials, but also, with the addition of new items to address parent and professional concerns over coverage and content validity, in a wider research context. Further studies are needed to assess its responsiveness to change.

Table 1 Reliability and floor/ceiling effects of the 4 ELDQOL subscales

ELDQOL Subscale	Cronbach's alpha	Item-total correlations	Intraclass correlation (test-retest)	Floor / ceiling effects (%)
Behaviour	0.79	.24 - .70	0.80	0 / 0
Seizure Severity	0.74	.19 - .54	0.96	2 / 0
Mood	0.86	.21 - .78	0.91	0 / 0
Side-effects	0.95	.33 - .88	0.92	10 / 0

Table 2 Validity: correlation between the 4 ELDQOL sub-scales and ABC and GHQ sub-scales¹

ELDQOL subscale:	ABC subscales		CHQ subscales		
	Irritability	Hyperactivity	Impact on Parental Time	Emotional Impact on Parent	Family Activities
Behaviour	r=0.008 p=0.95	r=0.13 p=0.32	r=0.25 p=0.1	r=-0.42 p<0.01	r=-0.31 p<0.05
Seizure Severity	r=0.29 p<0.05	r=0.38 p<0.01	r=-0.33 p<0.05	r=-0.47 p<0.01	r=-0.48 p<0.01
Mood	r=0.74 p<0.001	r=0.56 p<0.001	r=-0.28 p=0.06	r=-0.48 p<0.01	r=-0.63 p<0.001
Side-effects	r=0.38 p<0.02	r=0.39 p=0.01	r=-0.43 p<0.01	r=-0.54 p<0.001	r=-0.47 p<0.01

¹ Pearson's correlation coefficient

Table 3 Validity: mean scores of the 4 ELDQOL subscales by overall health

Overall health:				
	Very good/good	Fair/poor/v poor	t test	p value
Seizure severity	26	31	-1.8	0.08
Side-effects Profile	30	37	-1.9	0.06
Behaviour Scale	17	21	-3.6	0.001
Mood	32	35	-2.1	0.038

Table 4 Validity: mean scores of the 4 ELDQOL subscales by overall QOL

Overall QOL:				
	Very good/good	Fair/poor/v poor	t test	P value
Seizure severity	29	30	-0.2	0.81
Side-effects Profile	30	36	-1.4	0.16
Behaviour Scale	17	19	-1.8	0.078
Mood	32	35	-1.8	0.071

Table 5 Validity: mean scores of the 4 ELDQOL subscales by perceived severity of condition

	How severe:		t test	p value
	Very	Somewhat/moderate/ Mild		
Seizure severity	35	24	4.3	<.001
Side-effects Profile	43	32	2.9	<0.01
Behaviour Scale	21	17	3.1	<.01
Mood	37	31	2.4	<.03

Table 6 Validity: correlation between ELDQOL subscales and disability level

Scale	Pearson's <i>r</i>	p value
Seizure severity	-0.43	.001
Side Effects Profile	0.02	.93
Behaviour scale	0.44	.000
Mood	-0.1	.41

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